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A qualitative evaluation of the appropriateness, validity, acceptability, feasibility and interpretability of the Bristol Impact of Hypermobility (BloH) questionnaire

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We would like to acknowledge the assistance of the Hypermobility Syndromes Association in recruiting participants and to thank the interview participants for taking part in the research.

DECLARATION OF INTERESTS

There are no conflicts of interest.

HIGHLIGHTS

HIGHLIGHTS

- The BloH questionnaire was regarded positively by patients and physiotherapists.
- It is a welcome addition to the toolkit available to assess those with JHS.
- It captures the experiences of people with JHS.
- It does so in sufficient detail to support management.

ABSTRACT

BACKGROUND

The Bristol Impact of Hypermobility (BloH) questionnaire is a condition-specific patient-reported outcome measure developed for adults with Joint Hypermobility Syndrome (JHS). It has previously demonstrated strong concurrent validity with the Short-Form 36 health questionnaire and excellent test-retest reliability.

OBJECTIVES

This study aimed to evaluate its appropriateness, validity, acceptability, feasibility and interpretability.

DESIGN

A qualitative evaluation using semi-structured telephone interviews, incorporating a 'think aloud' exercise and additional prompts.

METHOD

Adults with JHS (n=11) were recruited through a patient organisation and physiotherapists with a professional interest in JHS (n=9) were recruited through the same organisation and an online professional network. Interviews were transcribed and data synthesised using a framework matrix.

FINDINGS

Patients and physiotherapists commented positively on the appropriateness, validity, acceptability and feasibility of the BloH questionnaire. Physiotherapists assessed the interpretability of the questionnaire and commented that, whilst further information might be captured, the value of that information might be limited. The questionnaire was considered comprehensive with only a very limited number of potential missing areas related to use of hand-held technology, hair washing/drying and intimacy. Interestingly, keyboard use and hair washing were excluded during initial questionnaire development as they were rated as relatively unimportant. Intimacy was not considered a comfortable addition for all participants.

CONCLUSIONS

Patients and physiotherapists regarded the BloH questionnaire as a welcome addition to the toolkit available to assess those with JHS. It was broadly accepted as

reflecting the experience of people with JHS in sufficient detail to support management.

KEYWORDS

Benign hypermobility syndrome; Ehlers-Danlos Syndrome,
Hypermobility Type; Qualitative Research; Questionnaire Design

MANUSCRIPT

INTRODUCTION

Joint Hypermobility Syndrome (JHS) is a heritable connective tissue disorder characterised by excessive range of movement in multiple joints and pain (Grahame, 2003). There is a lack of epidemiological data for adults in the general population, although the prevalence in musculoskeletal services is high, with 30% of referrals to one musculoskeletal triage service in the UK (Connelly et al., 2015) meeting the diagnostic criteria (Grahame et al., 2000). Similar high prevalence was reported in a UK rheumatology service (46% of women and 31% of men) (Grahame and Hakim, 2004); in women referred to musculoskeletal physiotherapy services in Oman (55%) (Clark et al., 2011); and in UK pain management (39.1%), rheumatology (37.0%) and orthopaedic lower limb clinics (10.9%) (To et al., 2017). JHS is associated with pain, fatigue, proprioception deficits and repeated cycles of injury (Terry et al., 2015). There is also evidence of anxiety and catastrophising (Terry et al., 2015), fear, agoraphobia, depression and panic disorders (Smith et al., 2014b). Its multisystemic nature is evidenced by associations with autonomic, cardiovascular, gastrointestinal,

gynaecological and urological conditions, and developmental coordination disorder/dyspraxia (Clark et al., 2014, Tinkle et al., 2017).

It should be noted that the diagnostic criteria and associated nosology have recently been revised, although the current research will use the term JHS as the study predated the new terms of 'hypermobile Ehlers-Danlos Syndrome (hEDS)' (Malfait et al., 2017) and 'Hypermobility Spectrum Disorders (HSD)' (Castori et al., 2017). Previous diagnoses of JHS and Ehlers-Danlos Syndrome Hypermobility Type (EDS-HT) were considered to be synonymous (Tinkle et al., 2009). A proportion, those with the most severe symptomatology, would now likely receive a diagnosis of hEDS (Malfait et al., 2017). The others would likely meet the criteria for 'generalised HSD' (gHSD) or 'historic HSD' (hHSD) (Castori et al., 2017). However, such diagnoses would need to be made on the basis of clinical examination by a clinician experienced in the assessment of connective tissue diseases (Malfait et al., 2017).

Physiotherapy is the main treatment for JHS, particularly exercise to enhance physical function. Two systematic reviews have, however, highlighted a lack of evidence to support therapy (Palmer et al., 2014, Smith et al., 2014a). A lack of congruence between the aims of physiotherapy and the outcome measures used to evaluate effectiveness has also been identified (Palmer et al., 2015). A condition-specific outcome measure has therefore been developed with patients to more accurately reflect the wide-ranging impact of JHS (Palmer et al., 2017a). The 'Bristol Impact of Hypermobility (BloH)' questionnaire gives a maximum score of 360, with higher scores representing more severe impact. It addresses items such as pain,

fatigue, physical function, anxiety, planning and management, and strength and weakness (Palmer et al., 2017a).

The BloH questionnaire has already undergone some evaluation of its psychometric properties. It correlates strongly with the physical component score of the Short Form 36 (SF-36) questionnaire ($r = -0.725$, $n = 615$) (Palmer et al., 2017a). High correlation coefficients were also observed for the majority of the physical component score subscales (physical function, role physical and bodily pain all $r > -0.7$, $p < 0.001$). The only SF-36 mental component score subscale that had a strong correlation with the BloH questionnaire values was social functioning ($r = -0.717$, $p < 0.001$). The overall mental component score and all other SF-36 subscales demonstrated statistically significant correlations (all $p < 0.001$) but with more moderate coefficient values ($r = -0.447$ to -0.624) (Palmer et al., 2017a). The BloH questionnaire has also demonstrated excellent test-retest reliability ($ICC = 0.923$, $n = 233$), performing better than the SF-36 in this regard ($ICC = 0.887$ and 0.778 for the physical and mental component scores respectively) (Palmer et al., 2017b). The smallest detectable change in the BloH score is 42 points, equivalent to a 19% change from baseline (as compared to a 25% and 37% change for the SF-36 physical and mental component scores respectively) (Palmer et al., 2017b). Finally the BloH has been shown to be more closely related to patients' global ratings of change ($r = -0.493$, $p < 0.001$, $n = 363$) than the SF-36 ($r = 0.186$ and 0.203 for the physical and mental component scores respectively, both $p < 0.001$ and $n = 363$) (Palmer et al., 2017b).

Important aspects related to validity, reliability and sensitivity to change have thus been established for the BloH questionnaire and it seems to perform very well in these aspects. However, other psychometric properties must be established before the BloH questionnaire can be confidently used to support JHS research and clinical practice.

This investigation therefore aimed to qualitatively evaluate specific properties of the BloH questionnaire, namely appropriateness, validity, acceptability, feasibility and interpretability. These concepts are defined in Table 1 and represent five of the eight criteria for evaluating patient-reported outcome measures for use in clinical trials (Fitzpatrick et al., 1998). Other criteria related to reliability, responsiveness and precision will continue to be addressed in future work.

METHODS

The study received a favourable opinion from the Faculty of Health & Applied Sciences Ethics Sub-Committee at the University of the West of England, Bristol (HAS/15/01/99).

Study design

Semi-structured telephone interviews using a 'think aloud' technique (also known as 'cognitive interviewing' (Drennan, 2003)) captured patients' and physiotherapists' views on the questionnaire.

Recruitment

Adults with JHS

Members of the Hypermobility Syndromes Association (HMSA), a United Kingdom (UK) patient organisation, were approached. Inclusion criteria were:

- ≥16 years old.
- Fulfil two or more hypermobility screening questions (Hakim and Grahame, 2003) and/or received a formal diagnosis of JHS or Ehlers-Danlos Syndrome Hypermobility Type (EDS-HT).
- No other formally diagnosed conditions affecting physical function.
- Able to give informed consent.
- Able to understand and communicate in English.

All criteria were self-declared. Recruitment packs were distributed to all HMSA members happy to be contacted about research (n=1 080). Packs included a participant information sheet, consent form, BloH and SF36 questionnaires (used for the associated test-retest reliability study (Palmer et al., 2017b)). Potential participants were separately asked to consider taking part in qualitative interviews, the results of which are the focus of this manuscript. The prospective target sample was n=10 which was considered sufficient to identify an appropriate range of opinions (Sandelowski, 1995). From the positive responses, patients were purposively selected on the basis of gender, age and BloH score.

Physiotherapists

Physiotherapists with an interest in JHS were also approached through the HMSA, who distributed recruitment packs (containing an information sheet and response slip) to all physiotherapists on their professional database (n=25). An invitation was

also placed on the interactive Chartered Society of Physiotherapy (iCSP) online professional network, with a request to contact the lead researcher ([initials of lead researcher]) for further information if they were interested. The prospective target sample was again $n=10$ (Sandelowski, 1995). It was anticipated that physiotherapists would be purposively selected by gender and years qualified however, due to the small number of responses, all were interviewed.

Interviews

Interviews were arranged by telephone and confirmed by e-mail. The aim was to learn what stakeholders thought about the BloH questionnaire. To achieve this, relevant 'manufactured data' (Silverman, 2013) was captured through interviews. Written informed consent was provided by participants in advance and verbal consent was confirmed at the beginning of each interview. A copy of the BloH questionnaire was sent to participants in preparation for the interview. The questionnaire is available as additional online material in the original report of its development and validation (Palmer et al., 2017a). Patient and physiotherapist interviews were undertaken concurrently. The interviewer (SM) was a very experienced postdoctoral researcher with extensive qualitative research experience, although this was her first involvement with JHS research. As such, she had no pre-conceptions.

Environment

All one-to one interviews were undertaken over the telephone (Silverman, 2013). The researcher used a quiet, private office using either a digital recorder or a hands-

free set (Taylor and Francis, 2013) with Audacity® software (version 2.1.1, Audacity Team, www.audacityteam.org, accessed July 2015) to capture voices.

A telephone interview proved supportive for patient participants. They sometimes identified themselves as fatigued or getting over a bad patch and, whilst happy to speak on the telephone, some adjustments were sometimes required (such as sitting in a particular chair or using the telephone on speaker mode to remove the need to hold it for long periods). However, such adjustments did not diminish their capacity to participate.

Physiotherapists often had academic or clinical commitments. Interviews were therefore accommodated at times such as before clinic or at lunch time. However the enthusiasm of physiotherapists to support the research mirrored that of patients.

Topic guides

Topic guides were informed by the researchers' tacit knowledge of appropriate literature and were reviewed by four experienced health professionals currently working with people with JHS (three physiotherapists and a rheumatologist).

Data sources

Three primary data sources were used:

- Retrospective 'think aloud' technique (Duncker, 1945) as a form of pre-testing by patients (Hagen et al., 2008).

- Further prompts aimed at eliciting specific information that might not have been captured on appropriateness, validity, acceptability, feasibility and interpretability (Fitzpatrick et al., 1998).
- Iterative interviewing was adopted to carry forward topics captured previously.

Transcriptions

Transcriptions of audio-recordings were anonymised and combined with prompts and researcher notes. Transcribers, independent from the research team, provided a non-verbatim/intelligent transcription, removing word repetition and false starts to sentences. The lead researcher then listened to the audio-recording whilst reading the transcripts to identify any errors or inaudible comments. Each transcript was cross-referenced with the notes made during and immediately after the interview. Interviews lasted 15 to 40 minutes. Member checking was not used as there is little evidence that it improves research findings (Thomas, 2017).

Data synthesis

The qualitative data was synthesised by the lead researcher using NVivo software (QSR International Pty Ltd. Version 10, 2012) and a framework analysis (Ritchie and Lewis, 2003). This enabled cross-sectional analysis against both the BloH questions and the relevant criteria for patient-reported outcome measures (Fitzpatrick et al., 1998). Data was coded against these largely a priori themes and criteria. Further criteria were identified which arose iteratively from subsequent interviews and these were added into the framework in an ongoing analysis. A matrix of both the patient and physiotherapist comments was then generated. The aim was to identify data relevant to the framework, rather than any notion of data saturation. Frequent review

and discussion with another research team member (SP) informed the ongoing refinement of the analysis framework and interpretation and ensured a rigorous analytical process.

FINDINGS

462 patient responses were received as part of the associated test-retest reliability study (Palmer et al., 2017b) (43% response rate), 348 of whom (75%) indicated that they were happy to take part in a telephone interview. From these, 11 were purposively selected. Patients were recruited from across the UK. Five had 'a paid job at present' (two full time and three part time), three were 'early retired due to sickness or disability', two were 'retired' and one was 'unemployed and seeking work'. All reported their ethnicity as 'White'. Patients scored a median of 4/5 on the hypermobility screening questionnaire (Hakim and Grahame, 2003). Nine physiotherapists responded and all took part in interviews. Participant characteristics are illustrated in Table 2. Reasons for non-response or non-participation were not sought. The findings are reported in relation to 'Appropriateness', 'Validity', 'Interpretability', 'Acceptability' and 'Feasibility' (Fitzpatrick et al., 1998). Table 3 provide illustrative quotes from participants. Other considerations that arose from the data are detailed subsequently.

'Appropriateness'

This criterion considers how well the content of an instrument matches the intended purpose (Fitzpatrick et al., 1998). We wanted to know how the BloH questionnaire reflected the experience of patients and if it captured sufficient detail. Across the

interviews both patients and physiotherapists commented positively on the appropriateness of the questionnaire content. A very small proportion of negative impressions related to the length of the questionnaire and potential repetitiveness but these were held in balance by the value of the content. The range of attendant symptoms such as fatigue was well received and participants felt that these inclusions provided a more holistic view of the effect of JHS. A small proportion of participants wished for increased focus on concomitant concerns, such as including *“autonomic issues, temperature control... brain fog, concentration issues”* [Patient 76] and *“I think it should have head in there and jaw”* [Patient 245]. Others reported co-morbidities that might have impacted on their answers to questions: *“Is it the POTS or is it the hypermobility syndrome?”* [Patient 33]. Reported co-morbidities included osteoporosis, irritable bowel syndrome, POTS, prolapsed rectum, chronic fatigue, headaches, and one patient was a wheelchair user.

‘Validity’

This criterion considered whether the questionnaire measured what it set out to measure (Fitzpatrick et al., 1998). The BloH questionnaire has a set purpose – to measure the impact of JHS on adults with the condition – and was being validated in relation to that purpose. Feedback identified how the questionnaire might be refined or improved in relation to measuring the effect of JHS in peoples’ lives in four areas:

Intimacy

Intimacy was flagged early by a physiotherapist for potential future inclusion:

“Um the only thing that was going through my mind is that - and I was just looking at C, the health and wellbeing section as well. There’s nothing that refers to um patient’s sexual activities and how they’re impacted by their mobility and their pain. So I know that’s an issue that is sometimes difficult for patients to talk about, but sometimes perhaps writing it down would be easier for them than bringing it up. A few, a few patients will bring that up with me, but obviously it’s a very difficult err issue to tackle and I sometimes read between the lines to get them to err talk about it um and discuss um issues to, to try to resolve some of the problems they have.” [Physiotherapist 2]

Tested iteratively with both groups, the addition of intimacy at some later point in questionnaire development was much welcomed by patients. Physiotherapists displayed surprise and, whilst they believed it might be useful, it appeared not to be a comfortable addition for them.

Section A of the BloH questionnaire

Section A asks *“During the past 7 days, have you had pain in any of the following areas?”* and requires a ‘Yes/No’ answer to ‘Shoulders’, ‘Elbows’, ‘Wrists’, ‘Hands’, ‘Hips’, ‘Knees’, ‘Ankles’, ‘Feet’, ‘Neck’ and ‘Back’. Almost half of the physiotherapists had no comments on Section A and read it without providing feedback. The majority felt that ‘head’ and to a lesser extent ‘jaw’ might usefully be added in the future.

There were conflicting comments as to whether there should be a more global non site-specific question around ‘pain’ as a type of ‘catch-all’.

Section E of the BloH questionnaire

Section E asks *“How much difficulty have you had with the following tasks during the past 7 days due to hypermobility?”* It was felt that the section would benefit from asking about the ability to use hand-held technology. There were also suggestions of other functional tasks that might be included, with the primary one being hair washing/drying.

“Um, washing hair’s a good one for people with fatigue because they find it really hard to wash their hair. You know, just lifting their arms up as a function, that’s often, that comes up quite a ... that’s ... that comes up quite a lot”

[Physiotherapist 8]

Interestingly questions related to computer use and hair drying had been incorporated at the first developmental stage of the BloH questionnaire (Palmer et al., 2017a) but both were removed from the final iteration as they were rated as relatively unimportant.

Section F of the BloH questionnaire

Section F asked *“How much discomfort would you have had after the following activities during the past 7 days?”* The section was broadly well received with patients commenting on its clarity:

“Um discomfort um there was absolutely nothing there that didn’t make sense um and wasn’t clear to answer.” [Patient 2]

“Section F was particularly useful on a personal level because it actually puts a time limit on activities that you can or can’t do as the case may be.” [Patient 11]

Some physiotherapists felt it was *“straight forward”* [Physiotherapist 7] or *“easy to answer”* [Physiotherapist 6].

Where a sub-section identified measuring discomfort after walking ‘a few miles’ or ‘wandering’, patients found this harder to judge. It was felt by some in both groups that it might be easier to provide a set time (such as 30 minutes or an hour) or set distance. However, there was not a strong steer that all would wish this to be altered. How these questions would be answered by a wheelchair user was raised by one patient who used a wheelchair on a daily basis.

‘Acceptability’

This criterion is concerned with the tool being acceptable to and for patients, minimising any distress, and obtaining high response rates (Fitzpatrick et al., 1998). Both groups indicated that the variety of responses provided enough opportunity and *‘variability’* [Patient 9] for the patient to complete, and were *‘easy to read and....easy to respond’* [Patient 10]. There were slight concerns about not using the same scales throughout but this was overshadowed by the broad acceptance that using a range of different response options would prevent questionnaire fatigue and promote engagement with the questions. All physiotherapists advised that that the range of responses were suitable. One physiotherapist commented that the variation of response options would slow people as they moved through the questionnaire but

another identified that the variation was helpful for completion. While one physiotherapist was concerned that patients might feel that this was a further questionnaire with little output, it was broadly well received.

'Feasibility'

Feasibility is related to the extent of effort, burden and disruption to staff and clinical care arising from use of an instrument (Fitzpatrick et al., 1998). Here, the issue of administering and processing the questionnaire was considered. There seemed to be little concern about this, although when it should be completed was queried (i.e. before or during an appointment):

"Um but I certainly think that if it was, you know, say I was with the consultant who'd got 20 minutes to get through all the patients and he's sat there filling this in I'd be screaming at him, 'Look at me. Get on with it.'" [Patient 6]

'Interpretability'

This criterion centres on how meaningful the scores are from an instrument (Fitzpatrick et al., 1998), and was only explored explicitly with physiotherapists. Through iterative work, additional areas arose that were continually tested for inclusion. However the physiotherapists commented that, whilst further information might be captured, its value might be limited in terms of what they could do to support patients with those aspects. As one physiotherapist commented:

"It's actually about what would be a um clinically significant difference, um yeah." [Physiotherapist 5]

The BloH questionnaire asks about symptoms in the last 7 days and it was evident that this required clarification as to why this cut off had been chosen:

“..giving some, yeah, a-appreciation to people why seven days has been chosen, because they’ll say, oh well I’m good this week but I’m, last week I was bad, you know?” [Physiotherapist 5]

“and, err, a lot of patients I see have episodic problems. So they might have had nothing in the last month, and then had a couple of months with a lot of pain.” [Physiotherapist 4]

“Seven days it’s err... I understand there might be a good reason for it, um but those symptoms come and go, they don’t... unless someone is very acute, um then they would obviously... did something that triggered err the symptoms or the osteoarthritis has kicked in so, so I don’t know, I’m not sure about the seven.” [Physiotherapist 1]

Patients with co-morbidities queried how these would be considered, as did some physiotherapists. For example, as reported in the section on ‘appropriateness’, a small number of patients had co-morbidities that might have affected how they answered questions. In a similar vein, both groups broadly felt that it would be useful to identify if patients had taken pain relief when the questionnaire was completed, as this would influence scores.

Guidance for completion is currently positioned at the top of Page 1 of the BloH questionnaire. However, the majority of participants from both groups moved directly to Section A during the 'think aloud' exercise and did not engage with the guidance. Unfortunately this was not explored further in the interviews.

DISCUSSION

The BloH questionnaire was broadly received positively by patients and physiotherapists. The development of a condition-specific questionnaire for JHS was welcomed as it offered validation to those patients who had perhaps been diagnosed later in their life, or felt that they had a range of symptoms that appeared disconnected. Patients often expressed gratitude for the research being undertaken, regardless of whether they were optimistic in outlook or weary that day, and they were happy to contribute.

The physiotherapists broadly felt that the BloH questionnaire would be acceptable to people with JHS. They indicated that patients would feel valued as the questionnaire explored the condition holistically, rather than as a cluster of unconnected issues. Physiotherapists identified that the BloH questionnaire would enable changes to be captured in more granular detail and that the range of symptoms forming patients' experiences of living with JHS were captured. The majority of physiotherapists identified the questionnaire as being useful to support patients and one was already using it in their clinic.

There were queries over the length of the questionnaire and whether its completion would adversely impact on appointment times, with a small number of both patients and physiotherapists suggesting completion outside clinic.

The lack of engagement with the 'Guidance for completion' instructions was unfortunately not explored further. It may be that both groups read the guidance when they first received the questionnaire and simply did not see the need to re-read it as part of the 'think aloud' exercise. In addition, sections A-H inclusive continually affirm a focus on the experiences of the last 7 days, thereby reconfirming much of the initial guidance for completion. This observation may not, therefore, be a significant issue. It was suggested that it might be helpful to ask those with dyslexia to assess the layout and typing of the questionnaire but this was not a major concern.

In terms of appropriateness, the BloH questionnaire appears broadly to measure what is of importance to patients. Fitzpatrick et al. (1998) commented that appropriateness is ultimately a judgement of fit between the questions and content of the tools. The fact that the questionnaire was developed with patient input from the outset (Palmer et al., 2017a) seemed to enable the patients to recognise their own experiences.

In terms of judging the validity of the BloH, another source of evidence is knowing how the questionnaire was developed in the first place (Guyatt and Cook, 1994). Extensive consultation was conducted with patients and two patient research partners throughout the development, refinement and initial validation of the

questionnaire (Palmer et al., 2017a). The value of this was identified by a patient in the current study who proffered that it was clear that the BloH questionnaire was developed by someone with the syndrome. Further evaluation by key stakeholders in the present investigation was aimed at ensuring further robust validation.

Regarding the criteria of interpretability, any changes in score would be of value where this meant that the management plan could be meaningfully amended. Both patients and physiotherapists felt that the questionnaire length should be balanced with the precision and value of the questions, their pertinence, clinical significance and how they informed management. What lay behind concerns expressed about the capture of pain relief and co-morbidities was that completion of the BloH questionnaire would vary depending on how patients were feeling generally. In separate ways, both groups spoke to the need to have a score that was meaningful and useful. Whilst one physiotherapist felt that the wording was too negative, in the main both patients and physiotherapists were positive about the acceptability of the questionnaire. It was seen to appropriately challenge those completing it by offering varied response options and preventing a 'tick-box' completion.

Strengths and Limitations

Data about the BloH questionnaire were captured from both those who live with JHS and physiotherapists with a particular interest in JHS. The sample of patients had a wide range of condition severity, with the median BloH score being very similar to the sample employed in the initial validation of the questionnaire (n=615, Palmer et al., 2017a) (BloH median score 238 versus 234 respectively). Although patients were overwhelmingly female, this is reflective of the population of those with JHS. Having

other formally diagnosed conditions affecting physical function was listed as an exclusion criterion but participants reported a range of co-morbidities and clearly did not self-exclude on the basis of common concomitant conditions. This enhances the validity of findings.

Participating physiotherapists were all female and all had many years of experience of working with people with JHS. No male or recently qualified physiotherapists commented on the BloH. The perspectives of other health professionals were also not included and these would be helpful to elicit in future research. Patients were recruited through a single patient organisation who might have an enhanced understanding of their condition (Terry et al., 2015) and they were slightly older than the cohort recruited for the initial validation of the questionnaire (Palmer et al., 2017a) (median 48 versus 39 years respectively). It is not suggested that the BloH is used with children since its development has been conducted exclusively with adults. It should be noted that participants self-declared a diagnosis of JHS/EDS-HT and, although this was supplemented by the five-point hypermobility questionnaire (Hakim and Grahame 2003), a clinical diagnosis cannot be assured.

As new diagnostic terms of hEDS and HSD are likely to include those with previous diagnoses of JHS or EDS-HT, the research team is confident that the BloH questionnaire will capture their experiences. However, this would need to be confirmed in future research. The ability of the questionnaire to discriminate between diagnostic categories would be another area for future research.

CONCLUSIONS

The BloH questionnaire was positively regarded by patients and physiotherapists alike. As a condition-specific outcome measure it was broadly accepted as reflecting the experience of patients with JHS and offers sufficient detail for therapists. This qualitative study has demonstrated that the BloH is a welcome addition to the toolkit available to support adults with JHS.

Ethical Approval: The study received a favourable opinion from the Faculty of Health & Applied Sciences Ethics Sub-Committee at the University of the West of England, Bristol (HAS/15/01/99).

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Conflict of Interest: There are no conflicts of interest.

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Table 1. Definitions of the criteria for evaluating patient-reported outcome measures explored in this research (Fitzpatrick et al., 1998).

Criteria	Definition
'Appropriateness'	Is the content of the instrument appropriate to the questions which the clinical trial is intended to address?
'Validity'	Does the instrument measure what it claims to measure?
'Acceptability'	Is the instrument acceptable to patients?
'Feasibility'	Is the instrument easy to administer and process?
'Interpretability'	How interpretable are the scores of an instrument?

Table 2. Characteristics of interviewees.

Patient	Sex	Age (years)	BloH Score (max 360)
1	Female	21	133
2	Female	50	108
3	Male	65	156
4	Female	83	170
5	Female	35	105
6	Male	48	265
7	Female	41	251
8	Female	35	238
9	Female	53	300
10	Female	31	300
11	Female	52	272
		Median (IQR) = 48 (35.0, 52.5)	Median (IQR) = 238 (144.5, 268.5)
Physiotherapist	Sex	Time since qualification (years)	
1	Female	30	
2	Female	37	
3	Female	28	
4	Female	37	
5	Female	30	
6	Female	37	
7	Female	27	
8	Female	31	
9	Female	12	
		Median (IQR) = 30 (28.5, 37.0)	

1 **Table 3. Illustrative quotes related to the criteria for evaluating patient-reported outcome measures (Fitzpatrick et al., 1998).**

‘APPROPRIATENESS’

“Yes, yeah, I think it’s fine.” [Patient 1]

“Um it, yeah it makes, it makes perfect sense to me.” [Patient 2]

“Well the only thing I can um... the downfall is, is over the last seven days, right. I mean we’ve had this for donkey’s years since I was 18, yeah.” [Patient 3]

“I think so, yeah. I think, yeah. I think it’s, err, it covers the, the breadth of, um, activities, so yeah.” [Patient 5]

“... you can tell that someone’s designed it who has got, you know, a real, er, understanding of the condition, which is great.” [Patient 7]

“... and it covers a lot of the symptoms that you feel that you might not realise that are part of hypermobility... I think it’s good because it actually makes you confront what you’re feeling, whereas if you go to anyone you put on, you know you have your face as it were. And this sort of opens you up a little bit to think well actually now hang on I do need help, and think to yourself well actually I do need some, do need help, and I do need a regime.” [Patient 8]

“The focus on hypermobility and how it impacts peoples’ ability to function on a day-to-day basis is fantastic... it captured all the main aspects of on a day to day basis it affects you.” [Patient 11]

“... it covers a great deal of the things that they discuss with me and that I um enquire about, so I think yeah it’s, it’s highly appropriate I would say... Yes, I do. I think it will be quite useful... So you’re talking things like breathing, you’re talking of the objective things but I do talk about sleep and fatigue and things like that and they go ‘no, I didn’t realise it was all associated, I thought it was just me.’ So yeah, no, I think it would, it can make them feel as though they’re being listened to and that’s a big thing.” [Physiotherapist 9]

“What’s good about it is that it does address issues that they commonly report, um so I think that’s helpful... think um it’s appropriate for people who are hypermobile; um I’ve just got some issues around the fact that there are so many co-morbidities, and this questionnaire doesn’t really um address those other bits.” [Physiotherapist 5]

“But overall, I think it’s great. I just think it’s a little bit wordy that was all and if we could, if you could consolidate it it would be great. Obviously, um, if you feel as though everything is in, I’m, I would really like to go ahead and have a go with a few patients and see how, how we got on.... Definitely [appropriate] (emphasis in interview)” [Physiotherapist 9]

‘VALIDITY’

“... it would certainly help um from a patient point of view me to help track err over time what’s going on.” [Patient 2]

“... I’ve been like this for so long I no longer know what the normal is. Um so I can say if I’ve felt frustrated with it, but then some of them say compared to your normal. I haven’t got a normal and I haven’t had for about 30 years.” [Patient 6]

“I think it would, it would help the patients feel validated as well because it’s asking you um really the things they can really struggle with, but... And when you ask them those questions yeah that then gives an opportunity to say, ‘Actually my life is out of control’, or ‘Actually I don’t get much help with this’, or ‘There’s no hope’, or whatever, you know.” [Physiotherapist 8]

“I think it covers a, um it covers a, a range of questions, so thinking about the specific joints it affects, pain, fatigue, activity. Um yeah I think, I think it’s, it’s quite comprehensive. I do... just some of it is a bit... wasn’t, wasn’t specific enough for my liking.” [Physiotherapist 8]

“Um I think there’s some useful, um very useful questions here um that cover some of the key areas that are big problems um around pain and fatigue, err just there’s this, there’s just an issue, I think, around some repetition. Um yeah.” [Physiotherapist 5]

Specific quotes related to intimacy:

“Now this is always a tricky one. I know particularly a lot of the girls with hypermobility have problems, well shall we say with hips dislocating at inconvenient moments for instance. I would think it could be useful because if you want a picture of where someone’s finding difficulty it’s no use just running away from an area.” [Patient 6]

“So I think it, even if it’s just a little question, you know, is it, is it affected, um, you know, do you struggle, that kind of thing would, would help. Um, and, and possibly, does your partner feel uncomfortable as well because a lot of the questions that come up, is my partner scared because he’s worried he’s going to put something out, like a joint.... And partners, partners do get worried that they’re going to break you.” [Patient 7]

“Oh yeah.. it’s no something you errr.. discuss freely with GP’s or rheumatologists but it is a major factor. ..it’s important and it is a major factor there is no doubt about it.” [Patient 11]

“Um, you know, I don’t ask so... But, but I, I, I, err, I - yeah, I have - I don’t have any strong feeling either way about it.” [Physiotherapist 4]

“Mm. I mean if you did it would just be um very um, you know, one question isn’t it, with like um does it affect your intimacy isn’t it or something. You know, yes or no isn’t it or five score or something, isn’t it, without going into any detail.” [Physiotherapist 8]

"It's not something that I usually go through unless somebody brings it up." [Physiotherapist 9]

Specific quotes related to Section A (body area):

"Um I have some headaches but it's not, not a biggie for me, but I can understand that it is for other people, so that would probably be useful, yeah." [Patient 2]

"Well that's an idea isn't it, yes. I hadn't thought of that because, um, it was, oh it must be a year or so ago I suppose, I yawned and for, um, well several weeks actually my, um, my jaw wasn't right, then it sort of slipped back you know." [Patient 4]

"Um.. yes, ...my oldest daughter has significant problems with jaw pain; she's been to ENT and they say it's related to her joint hypermobility, her mandibular joint." [Patient 11]

"I see that in question A, there is nothing about headaches, and that's quite a common problem I think in hypermobility. So, um, that would be, certainly be something I would want to record, err, on a form." [Physiotherapist 4]

"So the first thing I would think about with question A, as a clinician, is that, erm, it's interesting to see that 'head' isn't, isn't in the list because a number of the patients, erm, I have tend to have headaches or migraines, so that might be interesting to capture." [Physiotherapist 3]

'ACCEPTABILITY'

"But the questions are perfect. Um they are, they are, they seem like good questions... It's quite easy. Um the questions, the questions are easy to understand. I don't get confused by any of them. They're simple to follow." [Patient 1]

"Um I think there's a little bit of a usability thing around the, the scales that I was having an issue with." [Patient 2]

"Well initial, um it was, I thought it was quite long ((laughing)) and um, but, you know, it's um, the way it was formatted was okay. Some, some of them, you know, like had double meanings and, you know, you didn't quite... whether you should put one thing or another, but overall it was all right. You didn't have to think too much about it." [Patient 3]

"Um parts. Um I wouldn't say all of it because, for instance, the first part of it... that is very relevant to it because that makes me think, well exactly where has the pain been. That certainly is relevant and I can see and I can measure where it's changed, say, over a month there. Um some of the scale ones, they're where I struggle." [Patient 6]

"I mean, it covers, err, quite a lot. Some of it felt quite repetitive, um, at times. But, on saying that, it also because it was kind of repetitive but asked questions in a slightly different way, or trying to get a slightly different thing maybe, I could kind of understand that. Actually that's quite a good sense check to do and kind of a almost a double-check." [Patient 5]

"I do like the scoring from no discomfort, slightly uncomfortable, uncomfortable, painful, couldn't do it all. I thought that was really, really useful." [Patient 11]

"I would definitely um err put it in a more positive way. Um I, I found it very difficult... very um negative... I mean there is nothing about the, the physical activities they do and we... not much err psychological input in it either. Err often these people have anxieties... I think it's not holistic enough and it's, it's um, it's quite negative." [Physiotherapist 1]

"Yes I do, I do. Um it covers a great deal of the things that they discuss with me and that I um enquire about, so I think yeah it's, it's highly appropriate I would say." [Physiotherapist 2]

"Um, I think, well as I mentioned, I think that possibly for adults." [Physiotherapist 4]

"I think that will make people think and they won't just merrily tick along and not read the question." [Physiotherapist 9]

"Um yeah. I think um it's appropriate for people who are hypermobile; um I've just got some issues around the fact that there are so many co-morbidities, and this questionnaire doesn't really um address those other bits." [Physiotherapist 5]

"Yeah, you've mixed up, yeah, using numerical rating and um Likert. I think it's good to have variation because I think it does help with err questionnaire fatigue as well." [Physiotherapist 5]

"You know, I just, I just don't know, because what I get, you see, what I get from a lot of patients is that they are fed up with filling in lots of questions, lots of forms, scoring lots of things and getting all these measurements, and then nothing happening." [Physiotherapist 4]

'FEASIBILITY'

"Because at the moment this is just asking what's happened in the last seven days. Um, so, um, I would, um - if, if I only had information on what happened in the last seven days, I could only treat them on the basis of what has happened in the last - or manage them - on what has happened in the last seven days. Whereas if I knew what had happened before that, and that will be very valid and important in a hypermobile, um, then I would be able to base my, my management on that, or their management on that." [Physiotherapist 4]

"Well initially... if you were faced with four pieces of paper, although from my point of view I think great, you're asking all the right questions... from a patient's point of view, I think it needs to be a little bit more short and snappy... But overall, I think it's great. I just think it's a little bit wordy that was all and if we could, if you could consolidate it it would be great." [Physiotherapist 9]

'INTERPRETABILITY'

"It's going to get a clearer outcome because at the moment I don't have any form of functional outcome which I think is, is probably going to be, um, beneficial... Yeah. Um, well I think, I think what I, what it would do is it would be my baseline and then I'd do it as, um, it would probably make me look at whether I'd involve the occupational therapist from a fatigue point of view because they do a lot of fatigue here." [Physiotherapist 7]

"Yeah I think, yeah I think it would [inform management] wouldn't it, because then you'd, you'd pick up whether someone's feeling particularly weak or the pain's the main issue or they're actually not managing a lot of things like can't cope and... yeah. So you can... yeah you can be, you can sort of do your rehab plan around... because you'd... and sleep, and sleep's always a good one... I think it's quite good to have a subscale isn't it because then you can... after rehab or whatever you can compare the improvement can't you, you know?" [Physiotherapist 8]

"Um, it's going to be a challenge because the condition is so broad... how broad can one get, you know, for the higher functioning people um or the lower functioning people? It's getting, being able to capture the breadth of the condition um, you know, if you think about athletes or high level dancers, um and then at the other end of the scale um, you know, obviously those who are, who are very disabled. Um so it's kind of who's it going to be most appropriate for, or is there a ceiling or floor effect?" [Physiotherapist 5]

"I mean there is nothing about the, the physical activities they do and we... not much err psychological input in it either. Err often these people have anxieties. They... it's, it's err... so there is not much um of that as well." [Physiotherapist 1]